

GONOCOCCAL ULCERATION OF THE TONGUE IN THE GONOCOCCAL DERMATITIS SYNDROME*

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While a number of reports of cases of cutaneous gonorrhoea have been published, gonococcal manifestations in mucous membranes outside the genital tract and the conjunctivae seem to be very rare. Bronson (1919) collected twelve cases of gonococcal stomatitis from the literature. Only two of these cases were published later than the year 1909 when the differentiation between *Neisseria* species by fermentation was elaborated, and in only one case was it stated that the presence of gonococci had been proved by culture. Clinically, all presented as severe cases of diffuse ulcero-membranous stomatitis. Schmidt, Hjørring-Hansen, and Philipsen (1961) reported a similar though less severe case of presumed gonococcal stomatitis, probably contracted by cunnilingus, in a man with established gonococcal urethritis.

Diefenbach (1953) reported a case of proved gonococcal parotitis. Fiumara, Wise, and Many (1967) described an epidemic of gonorrhoea in a group of homosexuals. Among the fourteen patients were three with pharyngitis, two of whom were without clinical or bacteriological signs of urethritis or proctitis. One case of pharyngitis was definitely established as gonococcal, while fermentation tests failed in the other two owing to laboratory difficulties; even so the remaining data hardly leave room for reasonable doubt as to the gonococcal aetiology.

Bruusgaard and Thjøtta (1925) found haemorrhagic lesions in the oral cavity in their patient with gonococcaemia, gonococcal meningitis, and purpura. In one case of gonococcal dermatitis reported by Keil (1938), in which gonococci were grown from the blood, a few small vesicles surrounded by an erythematous halo were found on the soft palate. A case reported by Zachariae (1963) had multiple manifestations, including oral lesions, which were interpreted as allergic, arising after probable

eradication of the gonococci, being uninfluenced by penicillin, and healing after corticosteroid treatment.

Thus it seems of interest to report cases in which the gonococcal aetiology of oral lesions has been conclusively established.

The present communication describes a case of genital gonorrhoea complicated by ulceration of the tongue, dermatitis, and arthritis.

Case Report

A 39-year-old married woman employed as a domestic help was referred with an ulceration of the tongue, suggestive of primary syphilis.

Her past history was essentially negative, especially with regard to venereal disease.

She had noticed a sore on the tongue 5 days before referral and, at the same time, a rash on both hands and forearms. The following day there had been pain in the finger joints and general malaise, and in the next few days arthralgia developed in the wrist, elbow, and knee joints. She had had copious vaginal bleeding, but no chills, and she had not felt feverish.

Examination She was somewhat obese and did not appear acutely ill. Significant findings were restricted to the skin and tongue.

On the left margin of the tongue there was a fairly well-demarcated, dirty greyish coated, moderately tender ulceration measuring about 12 × 6 mm., with slight infiltration of the base (Fig. 1).

There were several small erythematous-papular lesions on the skin of the back and extremities, some with a central pustule, and on the forearms and hands were a number of larger vesiculo-bullous lesions, up to approximately 1 cm. in diameter, with a pronounced erythematous or violaceous border (Fig. 2). Only a single lesion was definitely haemorrhagic (Fig. 3), and there was no ulceration or crust formation anywhere.

The affected joints were somewhat tender, but without significant swelling, redness, or restriction of movement.

There was no lymphadenopathy and the temperature was normal.

Genital examination revealed inflammation of the skin and mucosa and a slight haemorrhagic secretion in the

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FIG. 1.—Ulceration of left margin of tongue from which gonococci were grown.



FIG. 2.—Vesiculo-bullous lesion in gonococcal dermatitis syndrome.



FIG. 3.—Haemorrhagic lesion.

vagina, but otherwise the appearances were unremarkable. No signs of early syphilis were found.

Laboratory Studies Repeated darkfield examinations from the ulcer on the tongue were negative for *Treponema pallidum*.

Smears from the tongue and skin lesions as well as

from the urethra and cervical canal were stained with methylene blue and by Gram's method. Numerous bacteria were found in material from the tongue and genital tract, including Gram-negative diplococci morphologically identical with the gonococcus.

Cultures from the same sites were examined at the State Serum Institute, Neisseria Department, and gonococci, identified by the typical fermentation pattern, were isolated from the urethra, cervix, and tongue. No gonococci were grown from the skin lesions, and repeated attempts at culture from fresh efflorescences on the following 3 days were also negative.

Sensitivity determinations showed the strain to be fully sensitive (50 per cent. inhibitory concentration of penicillin 0.0133 $\mu\text{g./ml.}$, of streptomycin <25.0 $\mu\text{g./ml.}$; and of tetracycline 0.20 $\mu\text{g./ml.}$ by the plate dilution method (Reyn, Bentzon, and Ericsson, 1963)).

The gonococcal complement-fixation test was positive, initially at a rather low titre (approx. 1:40), but repeat examinations showed rapidly and steeply rising titres, so that 10 days later it was very strongly positive (approx. 1:700).

Serological tests for syphilis were negative and remained so throughout the follow-up period.

The erythrocyte sedimentation rate was 35 mm. in the first hour, the blood picture was normal, streptococcal antibodies were slightly raised and the latex and Waler-Rose tests were negative. In the absence of pyrexia, blood cultivation was not attempted.

Biopsy of a bullous lesion on left thumb showed a sub-corneal bulla containing numerous leucocytes, but there was no infiltration of the epidermis. In the upper dermis there was a severe inflammatory reaction with leucocytic infiltration and histiocyte proliferation, and in several vessels there was pronounced endothelial hyperplasia with infiltration of the walls with leucocytes and lymphocytes; no thrombi were evident. No bacteria were found by Gram and Giemsa staining. The histological appearances were considered to be compatible with but not diagnostic of septic skin lesions produced by bacteria of low virulence.

Subsequent Course New skin lesions appeared daily, and malaise and migratory joint pains continued. The patient refused to enter hospital.

When positive results to gonococcal culture had been reported, the patient was treated with streptomycin, 2 g. intramuscularly in divided doses on the same day. All symptoms disappeared within 3 days, and the skin lesions dried up without scarring. The ulcer on the tongue healed more slowly, taking about a fortnight from the day of treatment. The titre of the gonococcal complement-fixation test fell considerably. Microscopical and cultural examinations for gonococci from the affected sites were negative 1 and 2 weeks after treatment.

Result Surveillance was terminated after 3 months, by which time the patient felt completely well, and showed no remnants of lesions.

Contact Tracing

Three sexual partners were traced and examined in venereal disease clinics. With two of them, peno-oral contact had taken place, 1 and 3 months respectively before symptoms appeared. Contact had been made with the third partner 2 weeks before the appearance of symptoms. In none of these cases was gonorrhoea demonstrated.

Discussion

Two kinds of gonococcal skin lesions may be distinguished: one caused by direct infection with gonococcal secretions, the other by bacterial emboli in gonococcaemia. The former leads to ulceration or an abscess-like process, often with considerable tissue breakdown, and gonococci are readily demonstrable. Examples of this type are cases reported by Pugh (1935, Case 1), Lowry and Franks (1943), Sears (1947), Marmell (1952, two cases), Byers and Bradley (1953), and Landergren (1961).

The other kind of lesion is typically seen in the gonococcal dermatitis syndrome, a gonococcal sepsis characterized clinically by fever, arthritis, and dermatitis brought about by small infarctions. The lesions usually present as vesiculo-pustules, sometimes haemorrhagic; they are few in number and show a predilection for the extremities. Gonococci are either not demonstrable or detectable for only brief periods. Cases of this syndrome have been published by Bruusgaard and Thjøtta (1925), Keil (1938), Margolin (1943), Kvorning (1963), Abu-Nassar, Hill, Fred, and Yow (1963), Ackerman, Miller, and Shapiro (1965), Björnberg and Gisslén (1965), Fred, Eiband, Martincheck, and Yow (1965), and Danielsson and Michaëlsson (1966).

In a number of cases of cutaneous gonorrhoea, *Neisseria gonorrhoeae* has been conclusively demonstrated in cultures from blood (Bruusgaard and Thjøtta, 1925; Keil, 1938; Ackerman and others 1965; Björnberg and Gisslén, 1965), from synovial fluid (Keil, 1938), and from skin lesions, both primary and metastatic (Bruusgaard and Thjøtta, 1925; Pugh 1935; Keil, 1938; Lowry and Franks, 1943; Margolin, 1943; Sears, 1947; Marmell, 1952; Byers and Bradley, 1953; Landergren, 1961; Fred and others, 1965). Danielsson and Michaëlsson (1966) demonstrated gonococci by immunofluorescence in skin lesions from two of their three patients with the gonococcal dermatitis syndrome. Bacteria with the morphological characteristics of the gonococcus have been found in biopsies of skin lesions (Ackerman and others, 1965; Bruusgaard and Thjøtta, 1925). In several cases, bacteria re-

sembling gonococci have been seen in smears from skin eruptions. Most authors stress that the chances of demonstrating gonococci are greater the younger the efflorescence chosen.

Knowledge of the infectivity of the gonococcus for the oral mucous membrane is limited. Few cases of gonorrhoea of this region have been reported, and the earlier ones are of doubtful validity because they antedate the development of fermentation tests for distinguishing the various *Neisseria* species. A number of these species are found normally in the upper respiratory passages, and their presence is additionally suggested by the significantly greater incidence of positive results to gonococcal-complement fixation tests in patients with bronchitis compared with that in controls (Bang and Krag, 1942; Lange, Reyn, Bentzon, and Lind, 1966). Thus the demonstration of a *Neisseria* in an oral lesion, without cultural identification, is no proof of its aetiological role.

The cases of gonococcal stomatitis collected by Bronson (1919) mentioned earlier presented a clinically homogeneous picture. In the case of Schmidt and others (1961), gonococci were not grown from the oral lesion, probably because treatment had been started some 12 hours before the sample was taken. Bacteria with the morphology of the gonococcus were found microscopically.

These cases, together with the three cases of gonococcal pharyngitis reported by Fiumara and others (1967), may be comparable to primary cutaneous gonorrhoea, appearing with a reasonable incubation period and in some cases being the sole manifestation of gonococcal infection.

In cases reported by Bruusgaard and Thjøtta (1925) and Keil (1938), oral manifestations probably of a metastatic nature occurred simultaneously with the cutaneous lesions at a time when there was proven gonococcaemia.

In the case presented here both explanations for the pathogenesis of the tongue lesion might be tenable. Gonococci could have been available both from the surface and from the blood stream, but the close time relationship to the occurrence of dermatitis strongly favours a metastatic pathogenesis. As is usually the case in this syndrome, the genital gonorrhoea was asymptomatic and may have been present for a long time, offering no clue to the date of infection. According to the patient's statement, infection probably took place 1 to 3 months before the appearance of symptoms and signs. It is hardly credible that the oral lesion should have appeared after so long an incubation period, although fairly prolonged incubation times have been quoted for primary cutaneous gonorrhoea. The absence of

gonorrhoea in this woman's sexual partners does not rule out direct infection, because they may have been treated in the interval and not admitted it. Furthermore, the antibiotic sensitivity pattern of the strain makes it probable that self-medication with small doses of an antibiotic would have been curative.

The clinical picture in this case is otherwise characteristic of the gonococcal dermatitis syndrome and resembles that seen in a series of patients from this department (Kvorning, 1963). There was no pyrexia at the time of examination, but the fever in such cases may be intermittent and sometimes low-grade, and the patient may have been febrile before consultation.

As it is certain that gonococci can produce disease in the upper respiratory passages it is strange that such cases are not encountered more frequently. They may perhaps be overlooked by both patient and doctor as a trivial "cold" not requiring bacteriological investigation.

The possibility of temporary, probably brief, colonization of the oral cavity with gonococci, as suggested by Bronson (1919), should perhaps be considered in those cases in which proven gonococcal infection seems inexplicable from the history and the findings on examination of the sexual contacts. It might be wise to pay more attention to this possibility by questioning the patients in more detail about their sexual habits and by closer examination of the oral cavity, including bacteriological investigation.

Summary

A case is presented of a female patient with asymptomatic genital gonorrhoea, gonococcal dermatitis, and arthritis, and an ulceration of the tongue from which *Neisseria gonorrhoeae* was cultured.

The pathogenesis of the oral and cutaneous lesions is discussed.

The possibility that infection or colonization of the oral mucosa with gonococci plays a part in the transmission of gonorrhoea in some cases is mentioned, and it is suggested that increased attention be paid to this possibility by more detailed questioning of patients and more frequent bacteriological investigation of the mouth and upper respiratory tract.

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REFERENCES

- ABU-NASSAR, H., HILL, N., FRED, H. L., and YOW, E. M. (1963). *Arch. intern. Med.*, **112**, 731.
- ACKERMAN, A. B., MILLER, R. C., and SHAPIRO, L. (1965). *Arch. Derm. (Chicago)*, **91**, 227.
- BANG, O., and KRAG, P. (1942). *Ugeskr. Laeg.*, **104**, 406.
- BJÖRNBERG, A., and GISSLÉN, H. (1965). *Nord. Med.*, **73**, 338.
- BRONSON, F. R. (1919). *Amer. J. Urol.*, **15**, 59.
- BRUUSGAARD, E., and THJØTTA, T. (1925). *Acta dermat.-venereol. (Stockh.)*, **6**, 262.
- BYERS, J. L., and BRADLEY, D. F. (1953). *Arch. Derm. Syph. (Chicago)*, **68**, 503.
- DANIELSSON, D., and MICHAËLSSON, G. (1966). *Acta dermat.-venereol. (Stockh.)*, **46**, 257.
- DIEFENBACH, W. C. L. (1953). *Oral Surg.*, **6**, 974.
- FIUMARA, N. J., WISE, H. M., and MANY, M. (1967). *New Engl. J. Med.*, **276**, 1248.
- FRED, H. L., EIBAND, J. M., MARTINCHECK, L. A., and YOW, E. M. (1965). *Arch. intern. Med.*, **115**, 191.
- KEIL, H. (1938). *Quart. J. Med.*, **31**, 1.
- KVORNING, S. A. (1963). *Dan. med. Bull.*, **10**, 188.
- LANDERGREN, G. (1961). *Acta dermat.-venereol. (Stockh.)*, **41**, 320.
- LANGE, P. K., REYN, A., BENTZON, M. W., and LIND, I. (1966). *Ugeskr. Laeg.*, **128**, 409.
- LOWRY, E. C., and FRANKS, A. G. (1943). *Amer. J. Syph.*, **27**, 428.
- MARGOLIN, E. S. (1943). *Urol. cutan. Rev.*, **47**, 512.
- MARMELL, M. (1952). *Amer. J. Syph.*, **36**, 88.
- PUGH, W. S. (1935). *Clin. Med. Surg.*, **42**, 125.
- REYN, A., BENTZON, M. W., and ERICSSON, H. (1963). *Acta pathol. microbiol. scand.*, **57**, 235.
- SCHMIDT, H., HJØRTING-HANSEN, E., and PHILIPSEN, H. P. (1961). *Acta dermat.-venereol. (Stockh.)*, **41**, 324.
- SEARS, H. J. (1947). *Amer. J. Syph.*, **31**, 60.
- ZACHARIAE, H. (1963). *Ugeskr. Laeg.*, **125**, 63.

Ulcération de la langue dans le syndrome de dermatose gonococcique

RÉSUMÉ

On expose le cas d'une femme présentant une gonococcie génitale asymptomatique, une dermatose gonococcique, une arthrite et une ulcération de la langue dans laquelle *Neisseria gonorrhoeae* fut trouvé par culture.

La pathogénie de ces lésions buccales et cutanée est discutée.

On mentionne la possibilité d'infection ou de colonisation de la muqueuse buccale pouvant jouer un rôle dans la transmission de la gonococcie dans quelques cas et l'on suggère d'attacher plus d'attention à cette éventualité en interrogeant plus soigneusement les malades et en pratiquant des recherches bactériologiques plus fréquentes de la bouche et des voies respiratoires supérieures.